Elastofibroma dorsi: clinical experiences of 13 cases and review of the literature

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Abstract

Aim: Elastofibroma dorsi (ELD) is a rare soft tissue benign tumor of the chest wall. We aimed to investigate the epidemiological, clinical, paraclinical, and treatment aspects of ELD. In the period between January 2009 and March 2018, 13 patients who underwent surgery for ELD were retrospectively analyzed in our study. Material and Method: Data of 13 ELD patients who underwent surgery were obtained retrospectively from medical records. Clinical findings were evaluated in terms of radiological, pathological and long-term results and compared with literature data. Results: During the study period 13 cases were operated for elastofibroma dorsi. Mean age was 52,4 (36-68) and there were 12 females and 1 male patient. The tumor was located on the right side in 8 patients (61,5 %), on the left in 4 patients (30,7 %) and bilaterally in 1 patient (7,6 %). All patients had complaints of swelling and back pain developed during the shoulder movements. All patients underwent complete resections. The tumor size ranged from 5 to 12 cm. The mean length of hospital stay was 3,6 days (1-6 days) with a morbidity of 15,3 % (seroma observed in 2 patients). The mean follow-up was 29,3 months (4-76 months). In the follow-up period, clinical symptoms of all cases were declined and no recurrence was observed. Discussion: Elastofibroma dorsi is a rare pseudotumoral lesion of soft tissue. In symptomatic cases, surgical excision should be preferred. When encountering painful masses in the periscapular area in elderly female patients, ELD should be considered on diagnosis.

Keywords

Elastofibroma Dorsi; Soft Tissue Tumor; Treatment

DOI: 10.4328/JCAM.5973 Received: 19.07.2018 Accepted: 29.07.2018 Published Online: 30.07.2018 Printed: 01.11.2018 J Clin Anal Med 2018;9(6): 557-60
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Introduction

Elastofibroma dorsi (ELD) is a rarely seen tumor-like lesion with a tendency to grow slowly; it is often seen in the subcapsular region and characterized by fibroelastic proliferation. First described by Jarvi in 1961, this lesion is called elastofibroma dorsi due to its characteristic subscapular-infrascapular localization [1]. This lesion is often asymptomatic, and the symptomatic patients often present with increased pain during shoulder movement or chronic back pain [2,3]. The physiopathology is not exactly defined. It has been thought that the mechanical friction between the chest wall and scapula that develops after recurrent trauma plays a role in the development of these lesions [4]. Diagnostics include the ultrasonography (USG), computed tomography (CT) and magnetic resonance imaging (MRI) methods [5]. However, MRI findings are often sufficient for diagnosis [6]. The disease is frequently treated by the excision of the mass [7,8]. Total excision is necessary to prevent recurrence and to relieve pain [9]. Their histological features include increased proliferating, thick, dark eosinophilic, fibrillar and globular elastic fibers [3,10].

The purpose of our study is to investigate the epidemiological, clinical, paraclinical, and treatment aspects of ELD and to emphasize the importance of the surgery in symptomatic cases.

Material and Method

Thirteen patients who were operated for elastofibroma dorsi between January 2009 and March 2018 were included in the study. The preoperative, perioperative and postoperative data of the patients were retrospectively reviewed. The cases were evaluated in terms of age, sex, complaints, clinical features, localization, size of the lesions, preoperative diagnostic methods, treatment, histopathological features and postoperative follow-up. They were subsequently compared with literature findings. Ultrasound examination, thorax computed tomography and magnetic resonance imaging examinations were applied alone or in combination for preoperative diagnostic evaluation. In all cases, a parascapular parabolic incision was performed in the prone position for complete resection. Pathological evaluations were reported to be consistent with elastofibroma. Informed consent was obtained from all patients for inclusion in the scientific paper.

Results

Thirteen patients were treated for elastofibroma dorsi during the study period. Twelve of cases (92.3 %) were female, 1 (7.6 %) case was male, and the average age was 52,4 (38-68). In five cases (38,4%), there was a history of heavy labor, while the rest of the cases did not have heavy labor history or heavy sporting activities in their anamnesis records.

Elastofibroma was observed on the right side of 8 patients (61.5 %), on the left side of 4 patients (30,7 %), and bilaterally in 1 patient (7,6 %). Both lesions of the patient with bilateral lesions were subscapular. Eleven of the lesions were subscapular (78.5 %) and 3 were parascapular (21,4 %) (Table 1).

The physical examination of all patients revealed increased pain and palpable swelling with shoulder movement as a major clinical finding. In addition to these findings, there were discomfort and clicking noises, which became evident by shoulder movements in 2 cases, and shoulder motion restriction in 4 cases (Table 1).

Ultrasonography and thorax computerized tomography (CT) examinations were performed for all patients in the diagnostic process (Figure 1). Magnetic resonance imaging (MRI) examination was performed in addition to these examinations in seven patients to obtain more data and to clarify the diagnosis (Figure 2). One patient underwent needle biopsy for malignancy exclusion and the result was interpreted as a benign lipomatous neoplasm. In the other cases, no diagnostic biopsy was performed due to the fact that clinical examination and radiological evaluation were sufficient.

In all cases, chest wall lesions were diagnosed as 'benign soft tissue tumor' by frozen examination, and then, total excision of the lesions was provided by muscle conservation approach. During the operation, latissimus dorsi was first opened in parallel to its fibers direction, and the mass was reached. Rubbery, moving lesions in yellow and white color were observed, which were tightly held under the periosteum of the ribs and under the scapula. In all cases, the chest wall muscle and bone structures were preserved and marginal excision was applied to the masses, so that chest wall integrity and rigidity were preserved. After the excision of the masses, the hemovac drain was placed

Table 1. Patients' characteristics and outcomes

Case	Age	Gender	Localization	Symptom	Size of tm.	Recurrence	Follow-up
1	63	F	R/S	CLİCK + M/P	11x6x4 cm.	_	52 months
2	48	F	BİL/S	SMR + M/P	R: 9x6x3cm. L:8x7x4 cm.	_	64 months
3	47	F	R/S	CLİCK + M/P	6x5x3 cm.	_	76 months
4	52	F	R/S	M/P	7x5x3 cm.	_	28 months
5	68	F	L/S	M/P	12x7x4 cm.	_	15 months
6	55	F	L/P	M/P	5x5x3 cm.	_	20 months
7	43	F	R/S	SMR + M/P	9x5x3 cm.	_	14 months
8	58	F	R/P	M/P	8x6x4 cm.	_	18 months
9	47	F	R/S	SMR + M/P	9x6,5x4 cm	_	40 months
10	38	F	R/P	M/P	9x7x3 cm.	_	36 months
11	49	F	L/S	SMR + M/P	7x6x3 cm.	_	8 months
12	56	M	R/S	M/P	6x7x4 cm.	_	6 months
13	58	F	L/S	M/P	6x5x4 cm.	_	4 months

F: Female, M: Male, R: Right, L: Left, BİL: Bilateral, S: Subscapular, P: Parascapular, SMR: Shoulder movement restriction, M/P: Mass/Pain

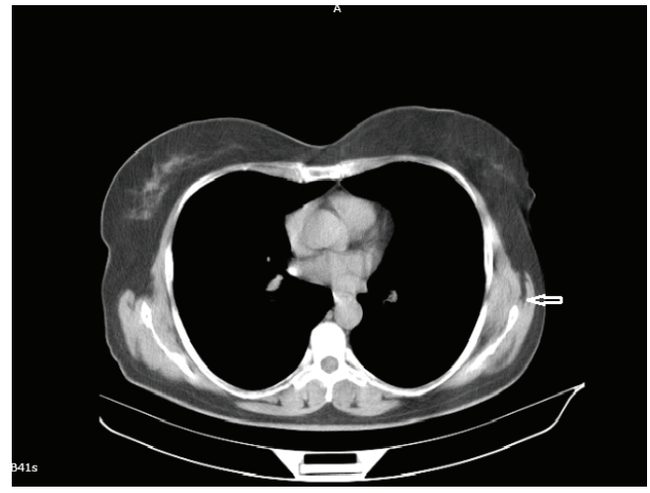

Figure 1. Computed tomography (CT) image of elastofibroma dorsi

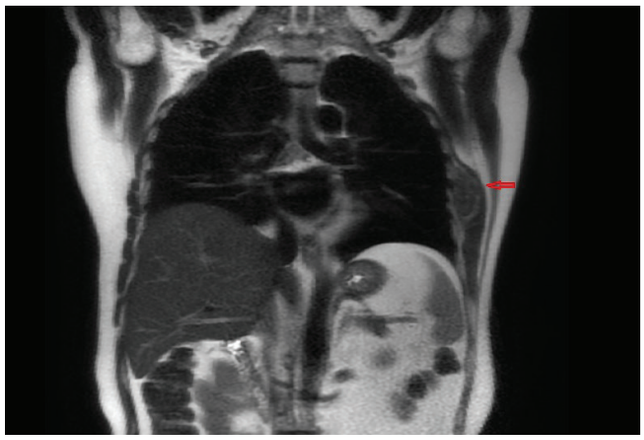

Figure 2. Magnetic resonance imaging (MRI) features of elastofibroma dorsi

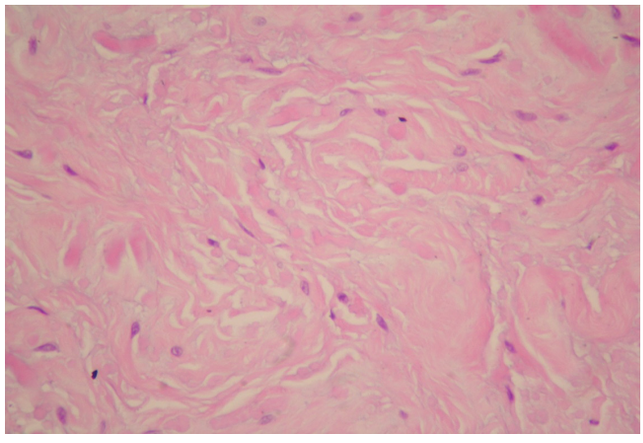

Figure 3. Bent or globoid elastic fibers and rare fibroblasts are visible in imaging degenerated with eosinophilic thick collagen (HEx400)

and the surgical layers were closed. The drains were removed 24-48 hours after the operation. The diameters of the resection material ranged from 5 cm to 12 cm. The mean hospital stay was 3.16 days (1-6 days).

Seroma developed in 2 cases (15.3%) in the early postoperative period. The cases could be treated without hospitalization through repeated needle aspirations and printed bandage treatments.

Pathological examination revealed macroscopically solid lesions on the surface of the cross-section with yellowish-white color, covered with fibrous capsules, and occasionally with fatty tissue. The hemotoxylin-eosin microscopic examination of the sections revealed mature adipocytes and generated elastic fibrils within the fibro-collagen tissue. The elastic fibrils were stained positive with the Elastica van Gieson staining kit (Figure 3).

The mean follow-up period of the cases was 29,3 months (4-76 months). In the follow-up period, clinical symptoms of all cases were in remission and no recurrence was observed.

Discussion

Elastofibroma dorsi is a rare, benign tumor of connective tissue that is localized in the chest wall, often located in the subcapsular region and which grows slowly and, has poorly defined borders [1]. Despite the fact that few cases related to this pathology were reached in the literature reviews, Brandser et al. have reported this rate to be 2% in their studies of thoracic CT in asymptomatic elderly populations [2]. It is usually seen in the population over 55 years of age, however, there are some cases that are detected at younger ages [3,4]. It was reported that it is seen 8-13 times more among women than in men [2,3]. The mean age in our study was 52,4 years, and 12 of the 13 ELD patients were female.

The frequency of bilateral occurrence of elastofibroma dorsi, which is usually observed unilaterally, is 10%. It is thought that bilateral cases are often synchronous [4,10]. More than 80% of elastofibroma dorsi are localized in the subscapular region, which is between the rhomboid and latissimus dorsi muscles and the sixth and eighth ribs. Sixty percent of the cases with subscapular localization are localized on the right [11]. Lesions localized in the tuber ischiadicum, foot, deltoid, axilla, trochanter major, olecranon, tricuspid valve, stomach, eye, hand, inguinal region and omentum are reported in the literature, however rarely [3,4,12]. In our study, ELD was bilateral in one patient (7,6%) and both lesions were subcapsular. In our study, 11 of the 14 ELD lesions were subcapsular, 7 of which were localized on the right (63,6%).

Elastofibroma dorsi pathogenesis cannot be explained clearly, but several important theories have been proposed. Firstly, Jarvi suggested that it was a reactionary condition, which was the result of excessive elastin production and collagen degeneration that developed due to the friction between the scapula and the thoracic wall [7]. This theory was supported by the fact that the condition was more common among people that do heavy labor with muscle power. However, it was later observed that the condition could develop among people that do not do heavy labor, and in different localizations. The later publications argued that reactive fibromatosis, degeneration due to vascular insufficiency, elastotic degeneration, enzyme defects and systemic involvement could play a role in the pathogenesis [3,8,11]. In addition, Nagamine et al. emphasized that there could be a genetic predisposition to etiology, because of the fact that they detected positive family history in 32% of the 170 cases in their study [3]. In our study, there was a history of heavy labor in the records of 5 cases (38,4%). With all these bases, it can be considered that the etiology of elastofibroma is multifactorial.

Asymptomatic cases make up more than half of the cases, and symptomatic patients usually present with increased pain and pain felt around the scapula [13]. In 90% of symptomatic cases, a mass lesion that is localized in the scapular region, and that became evident during shoulder movements, can be detected by physical examination [9,14]. In 25% of cases, a disturbing click may be felt due to shoulder movement [15]. Periscapular pain is seen in only 10% of cases [3]. It has been reported that the growth rate of these slowly growing lesions varies from 1 day to 67 years [3]. In our study, increased pain and palpable swelling with shoulder movement were observed in all cases. In addition to these findings, there were discomfort and clicking noises which became evident during shoulder movements in 2 cases (15,3 %), and shoulder movement restriction in 4 cases (30,7 %) (Table 1).

USG, CT and MRI examinations can be used for diagnosis of elastofibroma dorsi [5,16]. Chest graphs may detect soft tissue mass on the chest wall, but plain radiographs will not indicate specific findings regarding the characteristic of the lesion [16]. The most reliable imaging modality is accepted as MRI because the characteristic fibrous and fat component of the elastofi-

broma dorsi mass can be clearly recognized [5]. Diagnostic success was found to be lower than that of MRI, because CT is less sensitive to fatty tissue lineage within the mass [5,6]. Diagnostic radiological evaluation is mostly sufficient [6,13]. Needle aspiration or incisional biopsy may be performed to eliminate the possibility of malignancy. Despite the utility of needle biopsy in the diagnosis, it is not recommended because of the hypocellular nature of the lesion [17]. In our study, additional MRI was performed to clarify the diagnosis. In one case needle biopsy was performed for malignancy exclusion, and the result was interpreted as a benign lipomatous neoplasm. In the other cases, no diagnostic biopsy was performed due to the fact that clinical examination and radiological evaluation were sufficient. Hemangiomas, lipomas, desmoids tumors, neurofibromas, cicatricial fibroma, fibrous histiocytoma, fibromatosis, fibrolipoma and metastatic or primary sarcoma should be considered in differential diagnosis [17,18]. It is possible to observe mature fatty regions, fibroelastic cell proliferation, and eosinophilic collagen and elastic fibers in an interstitial mucoid ground microscopically [10,14]. Elastic fibers are typically in the form of small degenerate fragments like globules, discs and flowers [10]. Although immunohistochemical studies yielded different results; vimentin, actin, desmin and S-100 protein positivity were reported in neoplastic cells [10,19]. In these findings, elastofibroma has been suggested to originate from myofibroblastic and/or fibroblastic cells [19].

The recommended treatment for symptomatic cases or lesions larger than 5 cm. of elastofibroma dorsi is the excision of the mass with marginal borders [3,18]. On the contrary, it is suggested to avoid surgery especially in asymptomatic lesions that are smaller than 5 cm [8-14]. The most common complication after surgical excision is hematoma or seroma. For this reason, it is suggested that the bleeding control should be performed cautiously after excision of the mass, which is located in a large area [8]. Recurrence after total excision of the mass is not expected. Some cases of local recurrence have been reported in the literature, but it has been emphasized that recurrent lesions develop after incomplete excision [2,15]. No evidence of malignant transformation has been found in the disease, which has a quite long doubling time [20]. In our study, all cases were treated with marginal excision with the preservation of chest wall muscle and bone structures, and recurrence was not observed during follow-up periods.

As a result, elastofibroma dorsi should be considered when encountering painful periscapular masses in elderly women. The recommended treatment for these benign soft tissue tumors in symptomatic cases is total surgical excision.

Scientific Responsibility Statement

The authors declare that they are responsible for the article's scientific content including study design, data collection, analysis and interpretation, writing, some of the main line, or all of the preparation and scientific review of the contents and approval of the final version of the article.

Animal and human rights statement

All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. No animal or human studies were carried out by the authors for this article.

Funding: None

Conflict of interest

None of the authors received any type of financial support that could be considered potential conflict of interest regarding the manuscript or its submission.

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How to cite this article:
Şahin MF, Apaydın SM, Fındık G, Subaşı M, Yekeler E. Elastofibroma dorsi: clinical experiences of 13 cases and review of the literature. J Clin Anal Med 2018;9(6): 557-60.